



Available online at  
**ScienceDirect**  
[www.sciencedirect.com](http://www.sciencedirect.com)

Elsevier Masson France  
**EM|consulte**  
[www.em-consulte.com/en](http://www.em-consulte.com/en)



## Case report

# Bronchogenic cyst of the tip of the tongue: Report of two cases



J.-D. Kün-Darbois<sup>a,\*</sup>, R. Breheret<sup>b</sup>, A. Bizon<sup>b</sup>, A. Paré<sup>c</sup>, L. Laccourreye<sup>b</sup>

<sup>a</sup> Service de chirurgie maxillo-faciale et stomatologie, CHU d'Angers, 4, rue Larrey, 49100 Angers, France

<sup>b</sup> Service d'ORL et chirurgie cervico-faciale, CHU d'Angers, 4, rue Larrey, 49100 Angers, France

<sup>c</sup> Service de chirurgie maxillo-faciale et plastique de la face, CHU Trousseau, route de Loche, 37044 Tours cedex, France

## ARTICLE INFO

### Keywords:

Choristoma  
 Bronchogenic cyst  
 Lingual cyst  
 Tracheal bronchus  
 Congenital malformation

## ABSTRACT

**Introduction:** Lingual bronchogenic cyst is a rare congenital malformation, classified as a choristoma. It is lined by respiratory epithelium. Extrathoracic sites are rare and the tongue is an exceptional site.

**Case report:** The authors report two cases of bronchogenic cyst of the tip of the tongue in newborn infants. These two initially asymptomatic lesions gradually increased in size, leading to feeding difficulties in one case. MRI demonstrated well-demarcated, solitary, unilocular cystic images. Surgical resection was easily performed and histological examination concluded on bronchogenic cyst. An aberrant right upper lobe tracheal bronchus was also present in one case.

**Discussion:** Lingual choristomas are rare and defined histologically according to the type of epithelium. The embryological origin of lingual bronchogenic cysts remains controversial. MRI is the imaging examination of choice to characterize these lesions, but the definitive diagnosis is based on histological examination. Early, systematic surgical enucleation of all congenital lingual cysts must be performed to prevent obstructive and infectious complications.

© 2014 Elsevier Masson SAS. All rights reserved.

## 1. Introduction

Bronchogenic cyst is a rare congenital malformation. Lingual bronchogenic cyst constitutes a choristoma, a benign embryonic tumor characterized by the presence of normal tissue in an abnormal site (heterotopia). Bronchogenic cysts are lined by respiratory epithelium, i.e. ciliated, pseudostratified squamous epithelium, comprising mucus-secreting cells, smooth muscle cells and cartilaginous tissue. Extrathoracic bronchogenic cysts are rare and lingual sites are exceptional. We report on two cases of bronchogenic cyst of the tongue in newborns, associated with a tracheobronchial malformation in one case.

## 2. Case reports

### 2.1. Case 1

A nine-day-old boy presented with swelling of the tip of the tongue. Ultrasound revealed a unilocular cystic formation of the mobile tongue measuring 15 mm in diameter. MRI demonstrated

a cystic lesion with high-intensity signal on T2 and low-intensity signal on T1.

Needle aspirations were performed due to feeding difficulties and revealed a chocolate-coloured liquid containing well-differentiated squamous cells. Surgical resection was performed at the age of four months via a midline incision and submucosal dissection in contact with the cyst. A tracheal bronchus (anastomosis of the right upper lobe bronchus to the lateral surface of the trachea) was discovered during intraoperative fibroscopy performed for oxygenation difficulties. Histological examination concluded on bronchogenic cyst characterized by respiratory epithelium. A work-up looking for other malformations (echocardiography, transfontanelar and abdominal ultrasound, and full skeletal X-ray survey) was normal. The long-term course was uneventful with no signs of recurrence at 5 years.

### 2.2. Case 2

A three-month-old girl presented with swelling of the tip of the tongue. MRI revealed a midline, homogeneous, unilocular cystic lesion of the tip of the tongue, measuring 10 mm in diameter, with high-intensity signal on T2 and low-intensity signal on T1 with contrast enhancement after gadolinium infusion (**Fig. 1**). The lesion gradually increased in size. Surgical resection was performed at the age of four months via a midline incision on the dorsal surface of the tip of the tongue with dissection in contact with the cyst.

\* Corresponding author. Tel.: +33 6 62 00 88 34.

E-mail address: [jeandanielkun@yahoo.fr](mailto:jeandanielkun@yahoo.fr) (J.-D. Kün-Darbois).



Fig. 1. MRI (T1 + gadolinium) sagittal section revealing a lingual cystic image.

Histological examination concluded on bronchogenic cyst characterized by respiratory epithelium. The long-term course was uneventful with no signs of recurrence at 1 year.

### 3. Discussion

Lingual choristomas (or heterotopic cysts) comprise bronchogenic cysts and gastrointestinal duplications (lined by gastrointestinal epithelium) [1]. Cysts presenting both types of epithelium have also been described [2]. In the literature, these two types of cystic lesion are called choristomas, heterotopic cysts or foregut duplication cysts [1].

Lingual bronchogenic cysts are rare. In a review of the literature between 1942 and 1997, Manor et al. [3] described 52 lingual cysts, considered to be bronchogenic or gastrointestinal choristomas: 12 characterized by a respiratory epithelium (bronchogenic cysts), 25 characterized by gastrointestinal epithelium and 15 characterized by a combination of the two types of epithelium. In many articles the same terms were indifferently used to qualify bronchogenic cysts, gastrointestinal duplications or a combination of the two.

According to all authors, these cysts must be defined according to the respiratory or gastrointestinal nature of their lining epithelium [3].

To our knowledge, the association of bronchogenic cyst and tracheal bronchus has only been described once before, in the context of an intrathoracic bronchogenic cyst [4]. As no systematic review of this association has been performed, its frequency therefore remains unknown. It appears to be uncommon, although it is probably underestimated, as it is not systematically investigated. However, as this association does not have any clinical consequences, its diagnosis is probably unnecessary in routine clinical practice.

The aetiopathogenesis of these lesions has not yet been elucidated, but several hypotheses have been proposed in the literature. Bronchogenic cyst may result from an early aberrant nodule that becomes detached from the primitive tracheobronchial tree or may be derived from cells of the oesophagotracheal ridge, forming an isolated bronchial structure.

The usual site of these cysts is therefore the mediastinum or lung parenchyma, in which case the cyst may be adherent to or communicate with the tracheobronchial tree. Rarer, distant sites have also been described: intra-abdominal, cervical [5], cutaneous [6], intraoral.

As in the cases reported here, bronchogenic cysts are often discovered at birth, but they can also be asymptomatic and discovered later in adult life [7]. The main differential diagnoses for a congenital cyst of the tip of the tongue are choristomas (bronchogenic cyst, foregut duplication and bone choristomas), cystic lymphangioma, haemangioma, thyroglossal duct cyst, dermoid cyst or sialoceles [8]. The distinction between lymphangioma and these other diagnoses is important, as the first-line treatments are very different: injection of sclerosing agents into the lesion in the case of lymphangioma and primary surgical enucleation for choristomas, and even haemangioma and sialoceles. However, some authors also recommend first-line surgical treatment for lymphangioma due to the complications of medical treatment [9].

The differential diagnosis of ranula, although situated in the floor of the mouth, must also be considered due to its high prevalence and because it can be easily confused clinically with a lesion of the tip of the tongue.

When bronchogenic cysts increase in size, they can be complicated by upper airway obstruction, causing feeding and even respiratory difficulties and infection and possibly abscess formation [1]. Cases of malignant transformation have also been described in adults with untreated chronic lingual cysts, demonstrated to be bronchogenic cysts with malignant transformation to adenocarcinoma [10].

These lesions must therefore be surgically enucleated for diagnosis, treatment and prevention of possible complications. Surgical enucleation should be performed at the time of the first complications or at the age of one year, constituting a balance between the risks related to general anaesthesia, which decrease with age, and the risks related to possible complications.

### 4. Conclusion

MRI is the key examination in the presence of a cystic lesion of the tip of the tongue in children in order to evaluate the precise anatomical relations and confirm the unilocular nature of the cyst, which helps to guide the diagnosis. In this particular setting, bronchogenic cyst is the most likely diagnosis and sclerotherapy must be avoided. These lesions should be treated by surgical resection, allowing a definitive diagnosis. Like many authors, we recommend early surgical enucleation of all lingual cysts. This procedure must be performed from the first signs of complications or at the age of one year. No postoperative complications have been described in the literature and feeding can be resumed on the following day. However, like many authors, we recommend postoperative surveillance for 24 hours in order to rapidly detect any postoperative oedema.

### Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

### References

- [1] Blanchard M, Kadlub N, Boudjemaa S, et al. [Tongue cyst in children: foregut duplication, a possible diagnosis]. *Rev Stomatol Chir Maxillofac* 2012;113:442–7.
- [2] Joshi R, Cobb AR, Wilson P, et al. Lingual cyst lined by respiratory and gastric epithelium in a neonate. *Br J Oral Maxillofac Surg* 2013;51:173–5.
- [3] Manor Y, Buchner A, Peleg M, et al. Lingual cyst with respiratory epithelium: an entity of debatable histogenesis. *J Oral Maxillofac Surg* 1999;57:124–7.

- [4] Rodgers BM, Harman PK, Johnson AM. Bronchopulmonary foregut malformations. The spectrum of anomalies. *Ann Surg* 1986;203:517–24.
- [5] Borgnat F, Lupu Bratiloveanu P, Gyenes C, et al. [Bronchogenic cervical cyst in a child]. *Rev Stomatol Chir Maxillofac* 2011;112:54–6.
- [6] Kim PS, Cataletto M, Garnet DJ, et al. Unusual presentation of a cutaneous bronchogenic cyst in an asymptomatic neonate. *J Pediatr Surg* 2012;47:e9–12.
- [7] Petraud A, Khonsari RH, Corre P, et al. [Bronchogenic cyst of the tongue in an adult]. *Rev Stomatol Chir Maxillofac* 2009;111:46–8.
- [8] Akyol MU, Orhan D. Lingual tumors in infants: a case report and review of the literature. *Int J Pediatr Otorhinolaryngol* 2004;68:111–5.
- [9] Segado Arenas A, Flores Gonzalez JC, Rubio Quinones F, et al. [Severe iatrogenic airway obstruction due to lingual lymphangioma]. *Arch Pediatr* 2011;18:983–6.
- [10] Volchok J, Jaffer A, Cooper T, et al. Adenocarcinoma arising in a lingual foregut duplication cyst. *Arch Otolaryngol Head Neck Surg* 2007;133:717–9.